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Lethal *Strongyloides Stercoralis* hyperinfection in a young female with lupus nephritis; a rare case report

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Abstract

A young female, known case of lupus nephritis class III, on immunosuppressive drugs, presenting with continuous fever, diarrhea, cough, shortness of breath and hemoptysis. Physical examination revealed a non-healing ulcer on her right ankle with crepitation and wheezes heard bilaterally in her chest. Chest x-ray had bilateral infiltrates suggestive of interstitial lung disease while echocardiography, computerized tomography (CT) scan chest, bronchoalveolar lavage (BAL) and all cultures were negative. Antibiotics were started empirically but her condition did not improve. Later on, a palpable lymph node in the left axillary region was biopsied. In the meantime, the patient's condition had deteriorated and she died before the results of biopsy could be obtained. The lymph node biopsy revealed *Strongyloides stercoralis*-associated lymphadenitis. Thus, the biopsy resolved the complex clinical presentation of this patient with disseminated *S. stercoralis* infection.

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Introduction

Strongyloidiasis is a parasitic infection caused by the nematode, *Strongyloides stercoralis*. It is a worldwide problem, but mainly seen in the tropical regions of the world (1,2). We present a young female, known case of lupus nephritis class III, on immunosuppressive drugs, presenting with continuous fever, diarrhea, cough, shortness of breath and hemoptysis, who found to be infected with *S. stercoralis*-associated lymphadenitis.

Core tip

It is a necessity to keep a high index of suspicion for an early diagnosis of disseminated *Strongyloides stercoralis* infection in patients living in endemic areas for this worm and receiving prolonged immunosuppressive treatment.

Case Presentation

A 28-year-old female presented to us through the emergency department with continuous fever, diarrhea and a history of cough, hemoptysis and shortness of breath.

The patient was a known case of biopsy proven lupus nephritis, class III, and was on prednisone and cyclophosphamide treatment in standard doses.

On examination, a non-healing ulcer was seen on her right ankle; while bilateral wheezes and crepitation were present on her chest auscultation. Rest of her physical examination was normal. Bilateral infiltrates were seen on her chest x-ray, which were suggestive of an interstitial lung disease.

She was treated with antibiotics empirically but there was no response and the fever did not settle. During her workup, blood cultures, stool detailed report and culture and sensitivity and sputum for acid fast bacilli were found to be negative. Echocardiography, computerized tomography (CT) scan of chest and bronchoalveolar lavage (BAL) analysis were unremarkable. Her renal functions were also normal.

A few days later, a palpable left axillary lymph node was biopsied. In the meantime, the patient's condition worsened and she became drowsy and later on died before the findings of her lymph node biopsy report could be known. The lymph node biopsy revealed *S. stercoralis* infection (Figure 1).

Discussion

Strongyloidiasis is a parasitic infection caused by the nematode, *S. stercoralis*. It is a worldwide problem, but mainly seen in the tropical regions of the world (1-5).

The infection occurs in several forms, as primary infection, auto-infection, disseminated infection and has protean

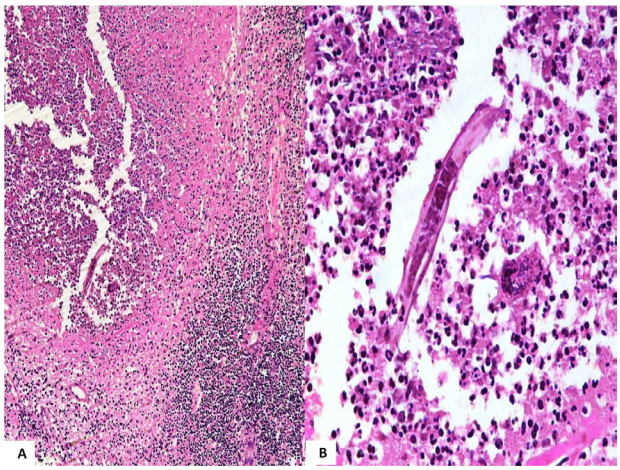

Figure 1. (A) Low-power photo showing large areas of acute suppurative and necrotizing inflammation involving the lymph node (H&E ×200). (B) Showing larval form of *Strongyloides stercoralis* surrounded by acute suppurative and necrotizing inflammation (H&E ×400).

manifestations from pauci-symptomatic to disseminated disease, the later known as *S. stercoralis* hyperinfection syndrome. Immunocompromised patients and those on immunosuppressive therapy are at particular risk for the development of *S. stercoralis* hyperinfection syndrome, which can prove fatal, if not diagnosed and treated early (2-4).

Systemic lupus erythematous (SLE) is an autoimmune disease that affects multiple organ systems. Since its treatment mainly revolves around steroids, this affects the immune system and leads to infections by opportunistic organisms. An example of such an infection, seen mainly in the tropics, is strongyloidiasis and is known as *S. stercoralis* hyperinfection syndrome (1).

During the primary infection, this parasite penetrates the skin to enter the human body, travels to the lungs, reaches the airways and is re-swallowed, finally ending up in the small bowel (1). The parasite can persist in the gut for life and can remain asymptomatic or produce a variety of symptoms.

The clinical presentation of *stercoralis* hyperinfection syndrome may range from gastrointestinal (GI) to pulmonary symptoms and the main factors predisposing to hyperinfection are the steroid therapy and human T lymphotropic viral (HTLV)-1 infections. Some associations with acquired immunodeficiency syndrome (AIDS), poor nutrition and malignancies has also been reported (2). In this respect, our patient was on prednisone and cyclophosphamide therapy for her renal disease.

Mora et al reported *stercoralis* hyperinfection syndrome in two patients with active SLE and antiphospholipid syndrome (APLS), one of which proved fatal (3). One of their patients was diagnosed on gastric biopsy while the other patient on BAL, investigation. We also performed BAL in view of respiratory symptoms and chest x-ray findings, but it was non-contributory in our case.

Arsić-Arsenijević et al reported a case of fatal *S. stercoralis* infection in a young female belonging to rural areas, and with lupus nephritis. Their patient presented with GI and pulmonary symptoms along with severe weight loss (4). Our patient also belonged to the same age group and background and presented with similar complains but weight loss was not observed in our patient.

Likewise, Sidoni et al reported a case of fatal *stercoralis* hyperinfection syndrome in a 78-year-old Italian male with temporal arteritis who was initiated on corticosteroid treatment 6 weeks ago. They recommended to screen the stool and other body fluids for parasites in patients starting on prolonged immunosuppressive therapy (5). However, this approach is not infallible as the chances of detecting infection on a single stool sample are only 25% (6). Stool detailed report and culture and sensitivity were both negative in our case.

Notably, an infection with this parasite can also lead to septic shock and organ failure (7). Potter el al reported *S. stercoralis* infection presenting as myocarditis (7).

Laboratory evaluation usually reveals an eosinophilia on complete blood count (8). However, eosinophilia may not be present and absence of eosinophilia leads to a delay in diagnosis. Similarly, eosinophilia was not evident in our case; probably due to immunosuppressive drugs.

Stercoralis hyperinfection syndrome can occur in patients even after anti-parasite treatment is started. Hence, it is necessary that the physician monitors the patients closely for the development of this complication (6). Treatment for this parasitic infection primarily consists of Ivermectin, with granulocyte colony stimulating factor (G-CSF) also being used as shown by Potter et al (7). Without treatment, death can occur (9). This was the case in our patient also. In our case, treatment could not be started as the patient expired before the diagnosis was made.

Conclusion

Our case shows how a patient on steroid therapy could not present with eosinophilia, a presentation that is often associated with infections by this parasite. And that a lymph node biopsy of any enlarged lymph node might also be useful in diagnosing disseminated *S. stercoralis* infection when other diagnostic methods have failed. Our case highlights the need to keep a high index of suspicion for an early diagnosis of disseminated *S. stercoralis* infection in patients living in endemic areas for this worm and receiving prolonged immunosuppressive treatment.

Authors' contribution

All authors contributed equally to the work.

Conflicts of interest

The authors declare no conflicts of interest.

Ethical considerations

Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors.

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